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Dentistry Section

Ameloblastic Fibro-odontoma or Immature Odontoma: A Retrospective Analysis of 134 Cases

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ABSTRACT

Introduction: According to 2017 WHO classification of odontogenic tumour, Ameloblastic Fibro-Odontoma (AFO) is no longer classified as an entity, probably representing immature stages of complex odontoma. However, there were few studies that revealed the differences between the lesion previously designated as AFO and complex odontoma.

Aim: To critically analyse the clinical, radiographic features and behaviour of AFO.

Materials and Methods: Eligible criteria included publications from PubMed, Scopus and Google Scholar reporting cases of AFO from 1975 to June, 2019 with available clinical, radiologic, and histologic information to confirm the diagnosis. Demographic data, localisation, size, treatment approach, follow-up period and recurrence were included.

Results: Analysis of 134 cases (124 previously reported and 10 new cases). The patient's age ranged from 7 months to 31 years (mean 10.3 years). There were 72 (54.1%) males, with a male-to-female ratio of 1.2:1. The mandible was involved in 79 (59%) cases, and the mandible-to maxilla ratio was 1.43:1. Nearly 80% of the lesions located in the posterior region of the jaws, and 48.5% were in the posterior mandible. Radiographically, most of the lesions were unilocular (95%) and only 5% were multilocular. The majority was mixed radiolucent radiopaque, and 15.8% were radiolucent. Almost all lesions (91%) were associated with the crown of an unerupted tooth. The range of follow-up was 6 months to 25 years. There were five recurrences among 134 cases accounting for a recurrence rate of 5.6%.

Conclusion: According to 2017 WHO classification of odontogenic tumours, AFO was not considered as an entity and was included in odontoma. However, there are some discrepancies between AFO and odontoma especially regarding the biologic behaviour. Therefore, long term follow-up for cases previously designated as AFO is warranted.

Keywords: Ameloblastic fibro-odontoma, Immature odontoma, Mixed odontogenic tumours

INTRODUCTION

According to 2005, the World Health Organisation (WHO) classification of odontogenic tumours, ameloblastic fibro-odontoma (AFO) was a rare mixed odontogenic tumour consisting of odontogenic epithelium in the stroma of odontogenic ectomesenchyme with hard tissue formation [1]. AFO was defined as a tumour with the histopathologic features of ameloblastic fibroma in conjunction with the presence of dentin and enamel. The prevalence of AFO was 0-3.4% within odontogenic tumours among different regions [1]. However, it comprised about 7% of the odontogenic tumours in patients under the age of 16 years [2].

Prior to 1967, AFO was confused with a lesion of similar nomenclature, the ameloblastic odontoma [3]. In 1971, however, WHO suggested that this term was inappropriate because it encompassed two types of odontogenic tumours that shared different histology and biologic behaviour [4]. Furthermore, Hooker SP distinguished between the two and emphasised the more innocuous behaviour of the AFO [5].

Generally, AFO is seen in the first and second decades of life [1,2,6]. The peak age ranged from 8 to 12 years [1]. It is more common in male than female [2,7-9]. The tumour usually occurs in the posterior region of the jaws, especially in the posterior mandible [6,9].

The most common presentations of AFO are painless swelling and delayed tooth eruption in the affected region. Most of AFOs are associated with unerupted teeth [10]. The radiographic feature of AFO generally reveals a well-defined radiolucent area containing various degrees of radiopaque mass of irregular size and form [2,6]. The ratio of radiopaque to radiolucent areas differs from one lesion to another; sometimes the mineralised element in the tumour predominates, and the lesion may resemble an odontoma [11]. Differential diagnoses of AFO includes lesions containing mixed radiographic pattern such as Calcifying Epithelial Odontogenic Tumour (CEOT), Calcifying Odontogenic Cyst (COC), immature complex odontoma and Adenomatoid Odontogenic Tumour (AOT) [12]. Microscopically, the tumour consists of dental papilla containing strands of odontogenic epithelium and immature tooth structures, including enamel and dentin surrounded by a fibrous capsule [2].

AFO is neither an aggressive nor an invasive tumour. Therefore, enucleation is considered to be the treatment of choice [7]. Although AFO has low potential for recurrence, though few studies reported the recurrence rate of 6.8% and 7.4% [7,9]. Moreover, the malignant transformation of AFO has been reported [13,14]. Slootweg PJ, proposed that the data on age, site, and sex were consistent with the concept that the AFO was an immature complex odontoma, thereby indicating that AFO was a hamartoma [6]. According to 2017 WHO classification of odontogenic tumour, AFO is no longer classified as an entity, probably representing immature stages of complex odontoma in most instances [15]. Nevertheless, there were few studies that revealed the differences between the lesion previously designated as AFO and complex odontoma.

The purpose of the present study is to critically analyse the clinical, radiologic features and behaviour of AFOs based on case reports and case series published in the English-language literatures, and to add ten new cases. The study aimed to provide updated information on disparities of biologic behaviour between AFO and complex odontoma.

MATERIALS AND METHODS

A retrospective study on AFO was done which consisted of two groups of data. The first group was composed of ten cases diagnosed as AFO being collected from the pathological reports of

the Department of Oral Pathology, Faculty of Dentistry, Chulalongkorn University (CU) from January 1, 1974 to June 30, 2019.

The second group was obtained from review of English-language literatures that were searched for adequately documented cases of AFO published between January 1, 1974 and June 30, 2019 using PubMed, Medline, Scopus and Google Scholar. The following terms were used in the search strategies: ameloblastic fibro-odontoma or ameloblastic fibro-odontome or mixed odontogenic tumour or mixed radiographic pattern. For studies appearing to meet the search words, the full reports were obtained. References of publication were searched for additional cases. The cases included in this study need to have enough of demographic, clinical, radiographic and histologic features to confirm the diagnosis of AFO. The minimum follow-up period of 6 months was also the criteria. Not all data were available for all cases. Cases that were reported multiple times were recorded once. Cases that were diagnosed as Ameloblastic fibro-dentinoma (AFD) were omitted from the present study because in the 2005 WHO classification, they were considered to be a variant of ameloblastic fibroma. Ameloblastic odontoma (AO)/ Odontoameloblastoma (OA) were also excluded. Review articles studied on immunohistochemical, genetic expression and other in-vitro studies that did not show enough clinical, radiologic, and histologic information were not included.

To determine the localisation of AFO, incisors and canines were regarded as the anterior region, premolars, molars and ramus (in the mandible) and premolars molars and sinus (in the maxilla) were regarded as the posterior region. The study was approved by the Research Ethics Committee of the Faculty of Dentistry, Chulalongkorn University (HREC-DCU 2018-042).

RESULTS

In the present review, there were some studies that did not meet the criteria. Even though the articles entitled Ameloblastic fibro-odontoma, the studies by Pillai et al., Ulgur, and Ghandehari-Motlagh M et al., were not included because the histopathologic features of these studies did not represent AFO [16-18]. Two articles [19,20] were also excluded due to the fact that they did not provide enough of demographic, clinical, radiographic and histologic features to confirm the diagnosis. Moreover, six cases [21-26] could not be included because they revealed the histopathologic features of AFD. Finally, a total of 134 cases of AFO (124 from publications and ten new cases from author's files) were analysed [6-8,11,12,14,27-92]. The data of ten new cases were summarised in [Table/Fig-1].

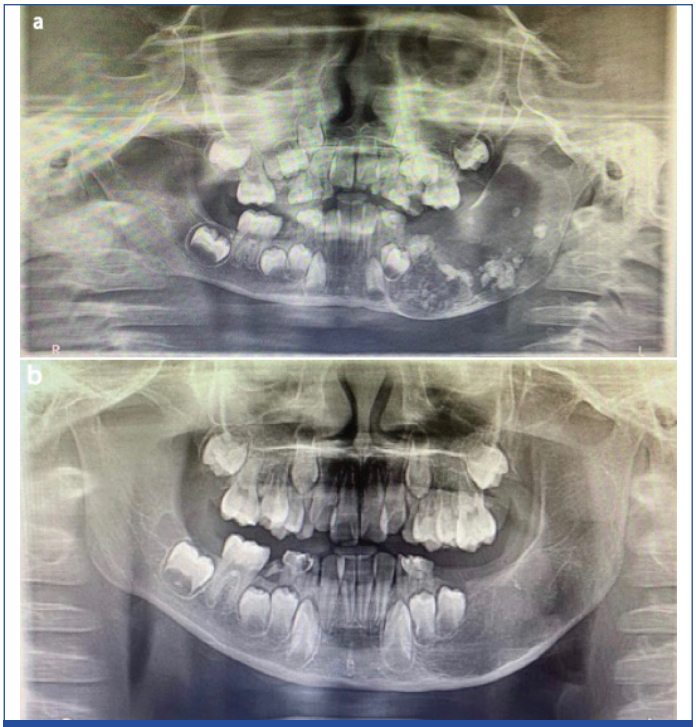
The age range of CU cases was from 2 years to 23 years with a mean age of 10.7 years. Seven cases (70%) occurred in male, male-to-female ratio was 2.3:1. Six cases (60%) were located in the posterior mandible. Six (60%) of ten tumours showed cortical bone expansion. Radiographically, eight (80%) of ten tumours were unilocular lesion. Eight tumours (80%) were radiolucent lesions with various amount of calcifications. Impacted tooth associated with the tumour was observed in nine cases (90%). All of the cases were treated by enucleation. Follow-up of the six lesions ranged from 5 to 14 years, and six of ten cases showed no recurrence.

The radiographic features of case 10 both before and 1 year after treatment were noted [Table/Fig-2a,b]. The histopathologic features of those ten cases disclosed the features of ameloblastic fibroma with varying amount of tooth-like structures. Obviously the soft tissue component dominated in case 5 and 7 with only small amount of tooth-like structures. Microscopy reveals nests and strands of odontogenic epithelium within primitive connective tissue resembling the dental papilla admixed with tooth structures consisting of dentin and enamel matrix [Table/Fig-3].

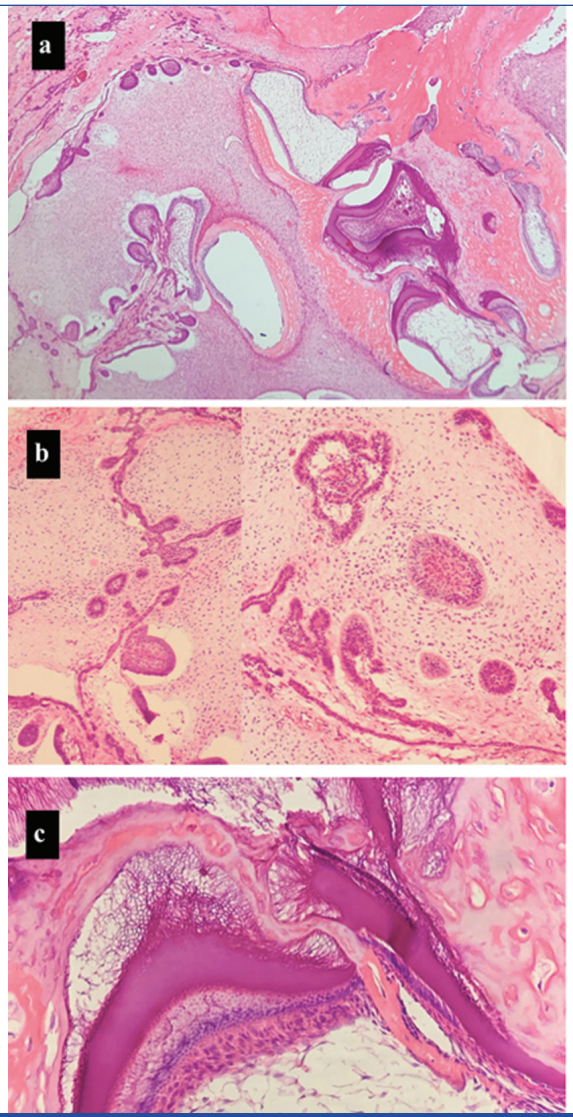
The clinical and radiological data of the 134 lesions were shown in [Table/Fig-4]. At the time of initial presentation, the patient's age ranged from 7 months to 31 years with an average age of 10.3 years. The majority of the cases were diagnosed in patients younger than 20 years (94.2%) [Table/Fig-5]. The majority (74.6%) of the lesions were located in the posterior region of the jaws, while 9% were located in both regions. The most common location was the posterior mandible in 65 cases (48.5%) [Table/Fig-6]. The clinical presentation of the AFOs was reported in 105 cases. AFO is characteristically painless swelling in 57 (54.3%) cases followed by failure of tooth eruption in 30 (28.6%) cases. In 18 (17.1%) cases, the lesion was asymptomatic and discovered incidentally on radiographic examination. Interestingly, there was a cortical bone perforation in 26 of 102 cases (25%). Almost all the lesions were unilocular lesions (95%), and multilocular lesions were uncommon (5%). Most of the lesions were described as radiolucent lesions with various amount of calcifications (82.2%) ranging from a few scattered opacities, a large mass of opacities and a single opaque mass (usually in the centre) that was surrounded by a narrow or wide area of radiolucency. Only a few cases showed unilocular radiolucency without calcification (15.8%). Most of the lesions in 112 (91%) cases were associated with an unerupted tooth, usually in the permanent dentition.

Case No.	Age (years)	Gender	Location	Clinical presentation	Expansion	Lobularity	Content	Impacted tooth	Clinical diagnosis	Follow-up period (years)	Recurrence
1	2	F	Ant max	Failure of tooth eruption	Yes	Uni	RP within well-defined RL border	Yes	CO	5	No
2	3	M	Ant max	Painless swelling	Yes	Uni	RP within well-defined RL border	Yes	CO	6	No
3	13	F	Post man	Swelling with infection	Yes	Uni	RP within well-defined RL border	Yes	CPO	11	No
4	9	M	Post man	Failure of tooth eruption	Not stated	Uni	RP within well-defined RL border	Yes	AF	14	No
5	10	M	Post man	Incidental finding	No	Multi	RL, No RP material	Yes	OM	Lost to follow-up	N/A
6	23	M	Post max	Incidental finding	Yes	Uni	RP within well-defined RL border	Yes	AO	Lost to follow-up	N/A
7	14	M	Post max	Painless swelling	Yes	Uni	RL, No RP material	Yes	AOT	Lost to follow-up	N/A
8	8	M	Post man	Failure of tooth eruption	No	Uni	RP within well-defined RL border	Yes	AF	9	No
9	17	F	Post man	Painless swelling	Not stated	Uni	RP within well-defined RL border	No	CPO	Lost to follow-up	N/A
10	8	M	Post man	Painless swelling	Yes	Multi	RP within well-defined RL border	Yes	AM	1	No

[Table/Fig-1]: Demographic and clinical features of 10 new cases of AFO.
M: Male; F: Female; Max: Maxilla; Mand: Mandible; Ant: Anterior; Post: Posterior; Uni: Unilocular; Multi: Multilocular; RP: Radiopaque; RL: Radiolucent; N/A: Not available; CO: Compound odontoma; CPO: Complex odontoma; AF: Ameloblastic fibroma; AO: Ameloblastic odontoma; AOT: Adenomatoid odontogenic tumor; AM: Ameloblastoma; OM: Odontogenic myxoma



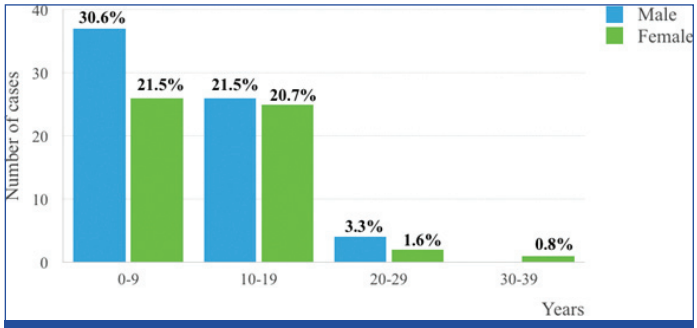
[Table/Fig-2]: a) Panoramic radiograph of Case 10 showing a well-defined multilocular radiolucency with scattered radiopaque foci resembling tooth-like structure on the left mandible; b) 1-year postoperative OPG reveals no evidence of recurrence.



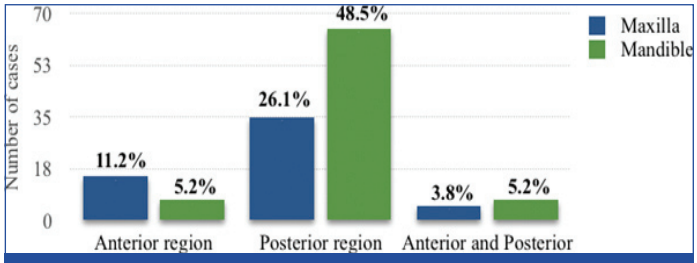
[Table/Fig-3]: a) Histopathological features of ameloblastic fibro-odontoma showing the soft tissue component and the hard tissue composed of disorganised tooth structure (H&E stain, 40X); b) The odontogenic epithelial cells arrange in narrow cords and small nests in a primitive connective tissue stroma resembling dental papilla (H&E stain, 100X); c) Dentin and enamel matrix are evident (H&E stain, 100X).

No. of cases	134
Age (year), mean	10.3
Gender, n (%)	133
Male	72 (54.1)
Female	61 (45.9)
Male-to-female ratio	1.2:1
Jaw,n (%)	134
Maxilla	55 (41)
Mandible	79 (59)
Clinical presentation, n (%)	105
Painless swelling	57 (54.3)
Failure of tooth eruption	30 (28.6)
Incidental finding by radiographic examination	18 (17.1)
Cortical bone perforation, n (%)	26/102 (25.5)
Radiographic features, n (%)	102
Unilocular	97 (95)
Multilocular	5 (5)
Tooth relationship, n (%)	123
Yes	112 (91)
No	11 (9)

[Table/Fig-4]: Demographic and clinical features of AFO in this study.



[Table/Fig-5]: Age of distribution of AFOs.



[Table/Fig-6]: Distribution of AFOs in the mandible and maxilla.

Follow-up information of 6 months or more were available for 89 cases. The range of follow-up was 6 months to 240 months. There were five recurrences establishing a recurrence rate of 5.6% [Table/Fig-7].

Author	Age (years)	Gender	Location	Diagnosis of Recurrent lesion	Recurrence	Follow-up
Howell RM and Burkes EJ, [14]	18	F	Post man	AFS	2 years	Patient died after multiple procedures
Furst I et al., [40]	7	M	Post man	AFO	2 years	6 months, NER
Friedrich RE et al., [58]	8	M	Post man	AFO	1.5 years	6 months, NER
Chen Y et al., [41]	2	M	Ant max	CPO	1 year	10 years, NER
	6	F	Post man	CPO	5 years	9 years, NER

[Table/Fig-7]: Recurrent AFOs.
M: Male; F: Female; Max: Maxilla; Mand: Mandible; Ant: Anterior; Post: Posterior; CPO: Complex Odontoma; AFO: Ameloblastic fibro-odontoma; AFS: Ameloblastic fibrosacroma; NER: No evidence of recurrence

DISCUSSION

AFO was previously recognised as an uncommon mixed odontogenic tumours. Slootweg PJ suggested that the classification of the AFO as a separate entity was superfluous and it should be regarded as an immature complex odontoma [6]. Cahn LR and Blum T, believed in "maturation theory", which suggested that AFO was an intermediate stage and eventually developed during the period of tooth formation to a complex odontoma thus, being a hamartoma [93]. On the contrary, other authors believed that AFO should be considered separately from odontoma since AFOs had a potential growth, leading to deformity and bone destruction therefore denoting the neoplastic nature [7,9]. Furthermore, malignant transformation of an AFO to an ameloblastic fibrosarcoma had been reported even though exceedingly rare [13,14]. Cases with malignant histomorphology, however, was not included in the study of Philipsen HP et al., and Slootweg PJ, [2,6]. Gardner DG, noted that some AFO were probably developing odontomas, whereas some were actual neoplasms [94]. Some authors reported that aggressive AFOs caused facial deformity, bone destruction and interfered surrounding structures [27-37]. In this review, six cases of AFO presented in patients over 22 years of age, which beyond the age of complete tooth formation. Surprisingly in this age group, AFO still existed, did not undergo maturation and became complex odontoma. Furthermore, the study revealed that 25.5% of cases associated with cortical bone perforation in line with 27% reported by Chrcanovic BR and Gomez RS, [9], in contrast to the clinical manifestation of odontoma usually showing absence of bone perforation. Hidalgo-Sánchez O et al., showed that the most common symptom of odontomas was retention of permanent teeth (57%) and the appearance of palpable tumour was less common (14%) [95].

The work of Chrcanovic BR et al., consisting of 215 AFOs were excluded because the study was undertaken without time or language restriction and included the articles before 1974, thereby did not meet the criteria used in this study [9]. Cases before 1974 were not included due to the fact that there was confusion in the nomenclatures of odontogenic mixed tumour.

The present study indicated that AFO was a tumour of children with the mean age of 10.3 years in accordance with the review by Philipsen HP et al., (9.0 years) and Boxberger NR et al., (9.4 years) and are more common in males than females (M:F ratio=1.2:1) [2,7]; similar to other publications [2,6,7-9]. According to the lesion location, this study showed that AFOs were common in the mandible (59%) especially in the posterior region (41%) in line with other studies [2,6-8].

The radiographic features of AFO generally revealed well-defined circumscribed unilocular radiolucency containing variable amounts of calcification more than multilocular appearance. The findings showed that only 5% were multilocular which was less than the study of Buchner A et al., (10%) [8], whereas odontomas seldom demonstrated multilocularity. Compare to complex odontoma, AFO possessed greater soft tissue component. When the amounts of the hard tissue component increased, complex odontoma had one mass of irregular calcification in the centre, while AFO had multiple scattered foci of dental hard tissue [96]. The 10 new cases revealed that the probability of correct clinical diagnosis of AFO was very low due to the fact that the lesion was uncommon. On the other hand, the radiographic features of odontoma are quite characteristic so it is easy to make a clinical diagnosis. Soluk Tekkesin M et al., claimed that the percentage of correct clinical diagnosis of odontoma by clinical and radiographic examinations was as high as 83.8% [97].

The presentation of a mixed radiolucent-radiopaque lesion of the jaws in patients younger than 20 years restricts the differential lists to a few odontogenic lesions. If the lesion locate in the anterior region of the jaws, AOT would be considered. However, the radiographic appearance of AOT reveals radiolucency containing fine snowflake calcifications. CEOT has a predilection for the posterior mandible and mostly occurs in the middle-aged patient. OA is an extremely rare lesion that contains ameloblastomatous component and odontoma-like elements. However, the latest WHO 2017 classification did not classify OA as a separate entity. There might be a difficulty in distinguishing between AFO and early stage of complex odontoma. In ten new cases from this study, there were variety of provisional diagnosis, only four (40%) of ten cases were diagnosed clinically as odontoma. The radiopacity of compound odontoma usually shows numerous tooth-like structures, whereas complex odontoma consists of a disorganised mass of calcified tissue. Since AFO is now included in odontoma, even in large radiolucency with small focal areas of calcification or without any calcification, it is also possible to make the differential diagnosis of odontoma in this kind of radiographic features. This review showed that a few AFOs were multilocular radiolucency, in such cases it is not usual to consider odontoma as a clinical diagnosis.

It was generally accepted that the AFO could be treated by enucleation. However, extensive lesions may require surgical resection [8,27,33,41,42]. A decision on whether or not to extract associated tooth/teeth remains with the treating clinician as success has been reported with either option. Even though unusually large size of complex odontomas were reported [95,97], however, they were not treated by surgical resection.

The present study showed a recurrence rate of 5.6% which considered less than that of Boxberger NR et al., 7.4% [7]. Three of the AFOs recurred within 1 to 2 years after the initial treatment. The reason for all five recurrences was supposed to incomplete surgical removal of the AFO from the first operation. In the recurrence reported by Furst I et al., there was an attempt to enucleate via curette preserving the associated tooth [40]. While Friedrich RE et al., excised the lesion including the germ of mandibular second molar [58]. However, Howell RM and Burkes EJ, reported one patient recurred with ameloblastic fibrosacroma 2 years after treatment and finally the patient died after multiple procedures [14]. According to Chen Y et al., two cases of AFO recurred as complex odontoma [41]. This finding supported the concept of progressive maturation that some AFOs were actually developing odontoma. As the case of malignant transformation existed in this review, thus the information supported the hypothesis that some AFOs may be neoplastic in nature.

According to 2017, WHO classification of odontogenic tumour, odontomas were subdivided into only two types: compound and complex odontoma, whereas AFO was not considered as an entity and was included in the group of odontoma. Interestingly, it is stated that the immature soft tissue counterpart containing cords and islands of odontogenic epithelium-a pattern similar to that seen in ameloblastic fibroma is regarded as the capsule [15]. However, this study revealed a few aspects of controversy between AFO and complex odontoma regarding the recurrence rate, the growth potential together with evidence of bone perforation and a very rare malignant transformation. Furthermore, some AFOs did not follow the concept of maturation theory as the data showed that they could recur as either complex odontoma or AFO. Therefore, should AFO be called an "immature odontoma" or classified as a variant of odontoma in order to differentiate it from the conventional complex odontoma. Besides, considering its immature nature, it may have a tendency to expand the alveolar bone more than that of mature odontoma, and as AFO consists of primitive tissue component "the dental papilla" so due to the fact that embryonal or primitive tissue is pluripotential and possesses the capacity to further differentiate thus leading to more aggressive clinical manifestations. Therefore, these findings confirm that the behaviour and prognosis of some AFOs are different from complex odontoma.

The present study showed a paradigm shift in establishing the differential diagnosis of odontoma, since this hamartoma also encompasses AFO. Besides, an understanding of the biologic behaviour of the various odontogenic tumours is of fundamental

importance to overall treatment of patients [98]. Therefore, long term follow-up for the cases previously designated as AFO is warranted. The possibility of recurrence should be under surveillance by the clinician.

LIMITATION

Complete records and data of clinical, radiographic features and post-operative records could not be accessed from all patients. Moreover, many cases had a short follow-up leading to underestimated actual recurrence rate. Molecular profile of AFO may reveal the correlation to its recurrence and biologic behaviour. Further analysis of genetic mutation of AFO comparing to odontoma would elucidate the molecular pathogenesis of the two lesions.

CONCLUSION

The present study revealed some discrepancies between previously designated as AFO and odontoma especially regarding to the biologic behaviour and recurrence. Thus, should AFO be called an "immature odontoma" or classified as a variant of odontoma in order to differentiate it from the conventional complex odontoma.

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